 doi: 10.2176/jns-nmc.2023-0263

NMC Case Report Journal 11, 79-84, 2024

A Ruptured Aneurysm of the Transdural Anastomotic Artery Occurring at the Temporal Base in Moyamoya Disease: Case Report and Literature Review

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Abstract

A 24-year-old woman with hemorrhagic onset moyamoya disease received bilateral indirect bypass surgery 11 years ago. She presented with a life-threatening atraumatic acute right subdural hematoma with temporal lobe intracerebral hemorrhage due to a transdural anastomosis (TDA) aneurysm rupture. We reviewed six cases of rare TDA aneurysms, all of which occurred around the temporal base. TDA aneurysms may occur near the main trunk of the middle meningeal artery (MMA) where hemodynamic stress is high. Therefore, we must pay attention to skull base aneurysms that form near the MMA; otherwise, the prognosis would be poor if such an aneurysm ruptured.

Keywords: transdural anastomosis, moyamoya disease, middle meningeal artery, subdural hematoma

Introduction

Moyamoya disease (MMD) is a chronic, occlusive cerebrovascular disease characterized by progressive vascular change.[1)] MMD usually causes some ischemic disease, and about half of adult patients with MMD develop intracranial bleeding. In bypass surgery, a new extracranial-intracranial (EC-IC; transdural) anastomosis is made to prevent ischemic events from occurring. Some authors have reported cases in which bypass surgery prevented the (recurrent) hemorrhagic event by the hemodynamic stress reduction of these fragile vessels or collateral vessels.[2,3)] Most bleeding sources are from ruptured, dilated fragile moyamoya vessels or dilated collateral vessels such as the anterior and posterior choroidal arteries.[4,5)] Additionally, transdural anastomosis (TDA) can also be a bleeding source because a TDA is architecturally vulnerable. Notably, TDA was the bleeding origin of subarachnoid hemorrhage (SAH) onset in patients with MMD where aneurysm was not confirmed.[6-8)]

However, an aneurysm arising from TDA, although rare, has been reported. TDA is common in MMD. It is formed by the periphery of the middle meningeal artery (MMA), occipital artery, internal maxillary artery, and ethmoid artery.[9)] The most frequent type of TDA is that formed by MMA, with a few case reports of the formation of an aneurysm and the development of an intracranial hemorrhage.[10-14)] Herein, we report the case of nontraumatic acute subdural hematoma (ASDH) with intracerebral hemorrhage (ICH) due to the rupture of a TDA aneurysm resulting in broad infarction with a poor outcome. We also reviewed reported literature discussing TDA aneurysm formation.

Case Report

A 24-year-old woman with MMD with severe consciousness disturbance was transferred to our department. Due to the onset of left thalamic hemorrhage, she underwent bilateral indirect revascularization (encephalo-duro-arterio-myo-pericranial synangiosis for the right hemisphere and encephalo-duro synangiosis for the left hemisphere) at our institution 11 years ago. She also underwent postoperative digital subtraction angiography (DSA) 10 years ago and showed good collateral angiogenesis on both cerebral hemispheres. We retrospectively confirmed the development of the right temporal fossa TDA (Fig. 1a-b, red cir-

Received November 6, 2023; Accepted January 17, 2024

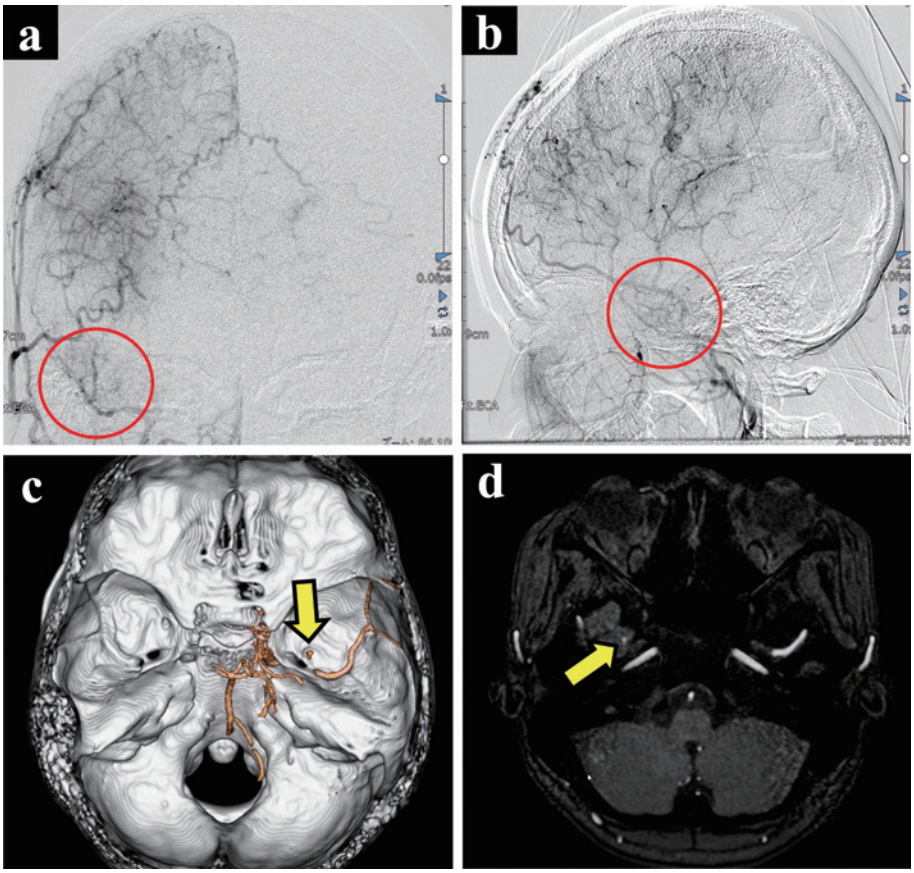

Fig. 1 Past digital subtraction angiography (DSA) and magnetic resonance angiography (MRA) fused with computed tomography (CT) scan.
DSA 10 years ago showed transdural anastomosis (TDA) from a middle meningeal artery (MMA) in the middle cranial fossa and staining in the same area (a–b, red circle area). MRA a year ago showed a TDA aneurysm in the middle cranial fossa (c–d, yellow arrow).

cle). However, the last follow-up MRI 1 year ago showed a small aneurysm in the middle cranial fossa; therefore, we decided to monitor this carefully because of the size (Fig. 1 c-d, arrow).

At emergent admission, the patient's consciousness score was 7 points on the Glasgow Coma Scale (E1 V1 M 5); her right pupil was 5 mm, and her left pupil was 3 mm. No external signs that indicated a traumatic event were found. Computed tomography (CT) showed a right ASDH with a midline shift and an atypical ICH at the bottom of the right temporal lobe (Fig. 2a-b). CT angiography (CTA) showed an aneurysm at the middle cranial fossa, which was thought to be the source of the bleeding (Fig. 2c-d, yellow arrow). We then performed emergent hematoma evacuation and decompressive craniotomy for lifesaving purposes.

Intraoperatively, we found a bleeding dilated vessel at the temporal base, which connected the dura and brain parenchyma (Fig. 3a). We then coagulated and cut this vessel; however, we could not find the aneurysm because it was buried in a thick hematoma. The right parietal lobes, considered to be perfused by indirect bypass, showed broad ischemic changes resembling a white-colored brain (Fig. 3b). After the operation, broad infarction occurred, and collateral blood flow from the external carotid artery system and the aneurysm disappeared, as shown in the CTA (Fig. 3c and d). She was finally transferred to another hospital with a modified Rankin scale score of 5 due to a disturbance of consciousness and without rebleeding.

Discussion

We present a rare case of a patient with a ruptured TDA-artery aneurysm occurring at the middle temporal skull base after an indirect bypass for MMD who suffered a life-threatening ASDH and ICH in the temporal base. Although an aneurysm was not confirmed by DSA in this case, it was determined to be an aneurysm because it was an arterial phase-specific intradural aneurysm-like structure located in the temporal skull base during the follow-up, and it was consistent with the bleeding point. In previously reported cases, MMD-related aneurysm was found at

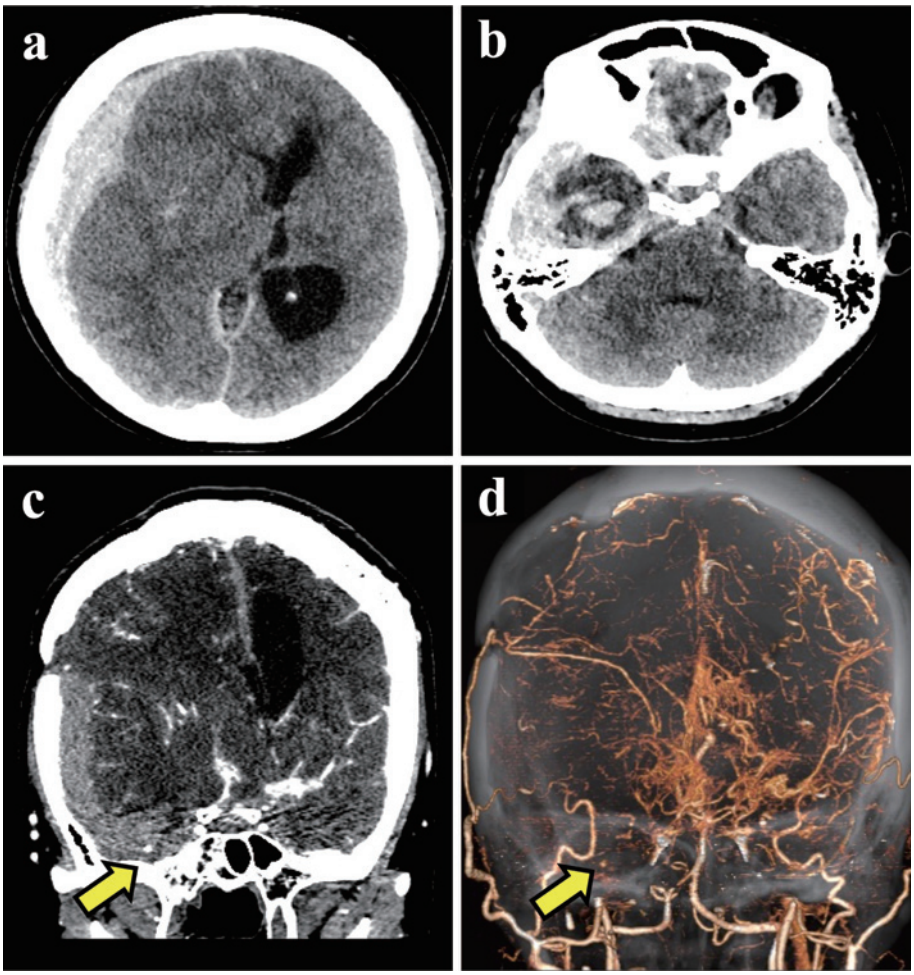

Fig. 2 CT scan and CT angiography (CTA) scan preoperatively.
CT revealed a right acute subdural hemorrhage (ASDH) with a midline shift (a). The primary site of the hematoma was in the middle cranial fossa (b). CTA and 3D-CTA showed the transdural anastomosis (TDA) aneurysm in the middle cranial fossa (c–d, yellow arrow).

a rate of 3.0%-12.0%, and most bleeding sources are dilated fragile moyamoya vessels or dilated parenchymal collateral vessels such as the anterior and posterior choroidal arteries.[4,5] However, the reported brain surface aneurysm arising from TDA is extremely rare, and there have been six cases of aneurysms arising from TDA, including the present case (Table 1).[10-14] We have reviewed six cases of TDA-artery aneurysm and of these, the patients in five cases were female (83.3%), and the patients in four cases did not receive bypass surgery and showed spontaneous well-developed TDA. Patients in four cases (66.7%) had an aneurysm near the MMA main trunk (near the foramen spinosum), and in two other cases, aneurysms arose from the posterior branch of the MMA, which was around the temporal skull base. Patients in five cases showed intracranial hemorrhage, and patients in two cases (33.3%) showed poor outcomes. Because this portion of the MMA and just branched TDA was directly affected by strong hemodynamic stress, its transdural collaterals from the MMA might easily form TDA aneurysm in this location (Fig. 3 e).[7,10] This may be the reason why the clearly identified TDA aneurysm is frequently found near the middle skull base but skull vault TDA aneurysm is not clearly identified. Some MMD cases were reported with faint SAH localized to the peripheral region, but these authors could not detect any aneurysm and discussed the presence of microaneurysm. According to this hypothesis, the size of TDA aneurysms might depend on the degree of hemodynamic stress of TDA. As temporal skull base TDA is not frequent, this aneurysm was considered to be rare. Unfortunately, we could not find any specific contributor for TDA formation in the temporal skull base. We speculated that previous bypass surgery might be an important factor of temporal skull base TDA, and only two cases (33.3%) of all six cases reported receiving a bypass surgery. The specific situation and mechanism that induce temporal skull base TDA is now unclear.

The outcome of our case was poor because a neurologi-

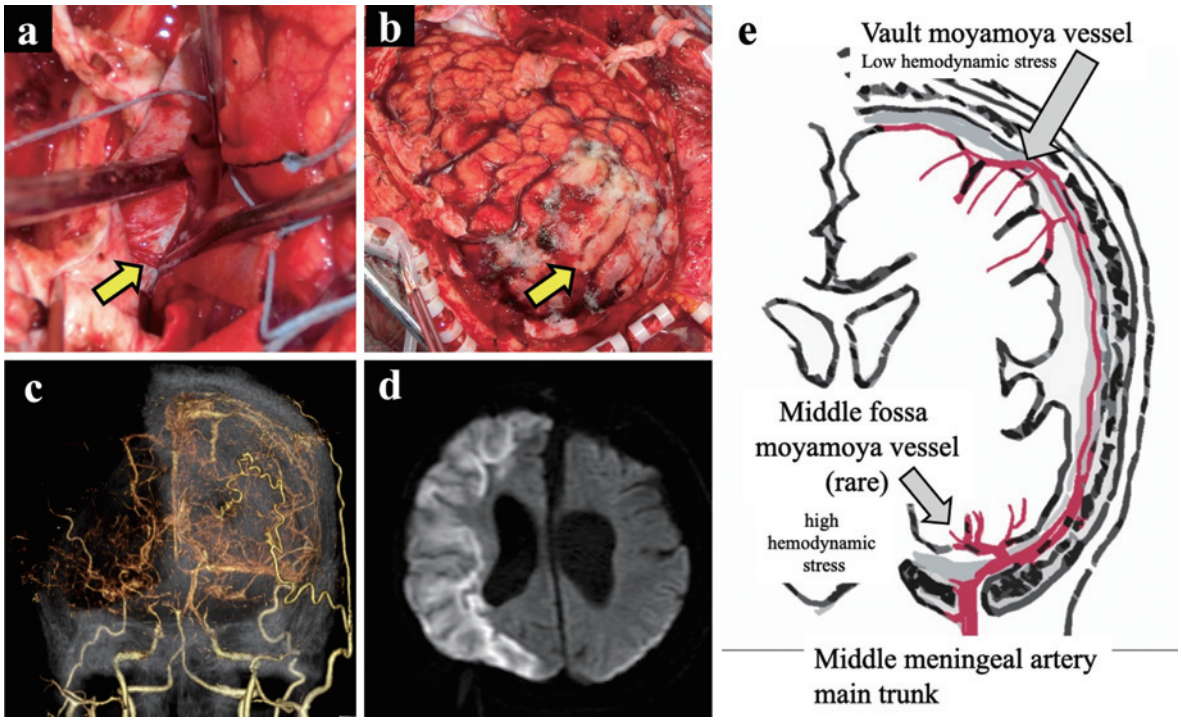

Fig. 3 Operative views and postoperative 3D-(computed tomography angiography) CTA scan and MRI and the mechanism of aneurysm formation in the transdural anastomotic artery.
We found a dilated middle meningeal artery (MMA) in the middle cranial fossa but did not find the aneurysm (a). The right parietal lobes where indirect bypass had been placed in the past had postoperative ischemic change (b, yellow arrow). Postoperative 3D-CTA showed no vascular abnormalities, including aneurysm (c). Postoperative MRI showed a broad infarction at the territory of the middle cerebral artery (d). Transdural collaterals from the MMA might easily form TDA aneurysm in the middle cranial fossa due to strong hemodynamic stress (e).

Table 1 Summary of transdural anastomosis artery aneurysms in patients with Moyamoya disease

Author and Year	Age/Sex	Previous History of Bypass surgery	Affected Side	Site of Aneurysm	Type of Bleeding	Outcome
Takahashi M[13] 1980	10/F	No	Left	Just distal of foramen spinosum and Parietal branch	No bleeding	NA
Borota L[10] 1996	41/M	No	Left	Posterior branch	SAH and ASDH	Poor
Koebbe CJ[11] 2004	31/F	No	Left	Posterior branch	Occipito-parietal ICH	Good
Park YS[12] 2010	43/F	No	Left	Just distal of foramen spinosum	SAH with temporal lobe ICH	Good
Umehara T[14] 2018	37/F	Right STA-MCA bypass	Left	Just distal of foramen spinosum	ASDH with temporal lobe ICH	Moderate
Present case	24/F	Bilateral indirect bypass	Right	Just distal of foramen spinosum	ASDH with temporal lobe ICH	Poor

STA: superficial temporal artery, MCA: middle cerebral artery, SAH: subarachnoid hemorrhage, ASDH: acute subdural hematoma, NA: not applicable
*Classified into good outcome, defined as a modified Rankin Scale score of 0-2, moderate as 3-4, and poor as 5-6, according to clinical sources.

cal condition was urgent, and we had to disconnect TDA arteries unavoidably to remove the hematoma. What is the best treatment in our case? For aneurysms located just distal of the foramen spinosum, it is difficult to identify the parent artery buried in the hematoma during emergent surgery. Alternatively, hemostatic treatment or aneurysm

embolization by endovascular therapy should be considered first if it is possible under the emergent situation.[15)]

In the present case, however, there was no time to perform endovascular therapy for a ruptured aneurysm because cerebral herniation caused by the ASDH and ICH occurred. Especially for the cases for whom indirect bypass has been performed in the past or the TDA was well-developed, it is necessary to preserve the TDA as much as possible.

In MMD, TDA poses a risk for MMA aneurysm formation, especially near the main trunk of the MMA, and serious intracranial hemorrhage, including ICH at the temporal lobe. Therefore, careful follow-up for the early detection of aneurysm formation and early treatment for the aneurysm near the skull base must be considered as soon as possible.

Conclusions

We report a case of a ruptured TDA aneurysm after an indirect bypass for MMD. This case report and literature review indicate that TDA at the skull base might contribute to MMA aneurysm formation, especially just distal of the foramen spinosum. Early detection and handling of aneurysms is important, as it is difficult to surgically treat aneurysms and preserve bypasses once a cerebral hemorrhage develops.

Acknowledgments

The authors thank Editage (www.editage.jp) for the English language editing.

Abbreviations

Moyamoya disease (MMD), extracranial-intracranial (EC-IC), transdural anastomosis (TDA), subarachnoid hemorrhage (SAH), middle meningeal artery (MMA), acute subdural hematoma (ASDH), intracerebral hemorrhage (ICH), encephalo-duro-arterio-myo-pericranial synangiosis (EDAMPS), encephalo-duro synangiosis (EDS), digital subtraction angiography (DSA), Glasgow Coma Scale (GCS), computed tomography (CT), computed tomography angiography (CTA), modified Rankin scale (mRS)

Statements and Declarations

All procedures performed were in accordance with the ethical standards of the institutions and the 1964 Helsinki declaration and its later amendments.

Consent to Publish

The participant has consented to the submission of the case report to the journal.

Author Contributions

YO: manuscript writing, figure composition, table composition, and manuscript drafting. JK: manuscript proofreading and image analysis. YN: manuscript proofreading. HY: manuscript proofreading. SK: manuscript proofreading. KS: manuscript proofreading. MM: manuscript correcting.

Funding

No funding was received for conducting this study.

Conflicts of Interest Disclosure

There are no conflicts of interest.

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